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Original Article

## LESIONS RELATED TO THE FORMATION OF BONE, CARTILAGE OR CEMENTUM ARISING IN THE ORAL AREA: A STATISTICAL STUDY AND REVIEW OF THE LITERATURE

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### Abstract

This report contains a statistical review of 559 cases of lesions forming hard tissues that were diagnosed by the departments of Clinical Pathophysiology and of Pathology at Tokyo Dental College from 1966 to 2001. Sixteen kinds of lesions which were related to the formation of bone, cartilage or cementum were analysed: osteoma, osteo-chondroma, chondroma, osteoid osteoma, osteoblastoma, ossifying fibroma, cemento-ossifying fibroma, cementifying fibroma, so-called cementoma, cementoblastoma, giantiform cementoma, periapical cemental dysplasia, osteosarcoma, chondrosarcoma, fibro-osseous lesion, and fibrous dysplasia of bone. The most common lesion was osteoma (203 cases). There is a marked tendency for this condition to occur in females (201 male cases and 358 female cases). The patients' ages ranged from 3 to 84 years, and the mean was 40.1 years old. Lesions with hard tissue formation were observed most frequently in the third decade and in the mandibular molar region.

Key words: Fibro-osseous lesion—Hard tissue formation—Bone—  
Cartilage—Cementum

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Part of the data (from 1966 to 1994) had been analysed and published in Japanese in *Shikwa Gakuho* (95: 545–556, 1995). New data from 1995 to 2001 were added and reanalyzed in this paper.

## INTRODUCTION

Although it is well known that many types of lesions involving hard tissue formation arise in the oral area, it is often hard to distinguish neoplasms from reactive lesions. The diagnostic decision determines the operative method employed and the controlling prognosis. For example, fibrous dysplasias of the bone are treated by orthopedic bone grinding, and ossifying fibromas are often excised. Therefore, this paper reports a statistical analysis and review of the literature about lesions related to the formation of bone, cartilage, or cementum arising in the oral area.

## MATERIALS AND METHODS

Out of a total of 32,625 cases seen by the departments of Clinical Pathophysiology and of Pathology at the Tokyo Dental College from 1966 to 2001, 559 cases of lesions involving the formation of bone, cartilage or cementum were reviewed. Sixteen kinds of lesions were considered in this study: 1. osteoma, 2. osteo-chondroma, 3. chondroma, 4. osteoid osteoma, 5. osteoblastoma, 6. ossifying fibroma, 7. cemento-ossifying fibroma, 8. cementifying fibroma, 9. cementoma, 10. cementoblastoma, 11. gigantiform cementoma, 12. periapical cemental dysplasia, 13. osteosarcoma, 14. chondrosarcoma, 15. fibro-osseous lesion, 16. fibrous dysplasia of bone. The numbers of each lesion and the frequency by gender, region and age were analyzed for each type of lesion. A huge lesion, for example, from the premolar to the molar region, was counted in both regions. Lesions arising in the periphery of gingiva or with dystrophic calcification were excluded from this study.

## RESULTS

### 1. Frequency of each lesion (Table 1)

The frequency of lesions involving hard tissue formation was 1.7% of all specimens

Table 1 Frequency of each lesion

Lesions	Number	Percentage
Osteoma	203	36.3
Osteo-chondroma	4	0.7
Chondroma	1	0.2
Osteoid osteoma	3	0.5
Osteoblastoma	4	0.7
Ossifying fibroma	60	10.7
Cemento-ossifying fibroma	25	4.5
Cementifying fibroma	49	8.8
Cementoma	38	6.8
Cementoblastoma	3	0.5
Gigantiform cementoma	7	1.3
Periapical cemental dysplasia	22	3.9
Osteosarcoma	16	2.9
Chondrosarcoma	3	0.5
Fibro-osseous lesion	65	11.6
Fibrous dysplasia of bone	56	10.0
Total	559	100.0

diagnosed by the departments of Clinical Pathophysiology and of Pathology of Tokyo Dental College. Table 1 shows the numbers and percentages of each type of lesion. Osteoma was the most common lesion; the second most frequent was ossifying fibroma. Only one case of chondroma was diagnosed.

### 2. Frequency by gender (Table 2)

Table 2 shows the numbers of lesions involving hard tissue formation according to gender: they arose on females 1.78 times more often than on males, *i.e.* 201 cases (36.0%) were males and 358 cases (64.0%) were females.

### 3. Frequency in each region (Table 3)

Lesions with hard tissue formation arose in various regions of the oral cavity and around the oral area. The most common region in which such lesions were found was the molar region containing the maxillar and mandibular areas. The percentage arising from the mandibular molar region was 32.2%, and that arising from the maxillar molar region was 15.6%. The frequencies in other areas were 41 cases (5.4%), including 16 cases of palate (sixteen osteomas), 8 cases of the man-

dibular angle (4 fibrous dysplasias of the bone, 2 osteomas, 1 ossifying fibroma and 1 fibro-osseous lesion), 4 cases of the mandibular ramus (2 fibrous dysplasias of the bone, 1 osteoma and 1 osteoblastoma), 4 cases of the

maxillary sinus (4 osteomas), 4 cases of the tongue (3 osteomas and 1 osteo-chondroma), 2 cases of the mandibular condyle (1 osteoma and 1 osteo-chondroma), 1 case of the mandibular plane (an osteoma), 1 case of the temporal region (an osteoma), and 1 case of the nasal cavity (an osteosarcoma).

Table 2 Frequency by gender

Lesions	Male	Female
Osteoma	90	113
Osteo-chondroma	2	2
Chondroma	1	0
Osteoid osteoma	2	1
Osteoblastoma	2	2
Ossifying fibroma	20	40
Cemento-ossifying fibroma	3	22
Cementifying fibroma	12	37
Cementoma	18	20
Cementoblastoma	1	2
Gigantiform cementoma	2	5
Periapical cemental dysplasia	5	17
Osteosarcoma	8	8
Chondrosarcoma	1	2
Fibro-osseous lesion	13	52
Fibrous dysplasia of bone	21	35
Total	201	358
Percentage	36.0	64.0

#### 4. Frequency by age (Tables 4, 5)

The patients' ages ranged from 3 to 84 years; the mean was 40.1 years old. The youngest patients were two 3-year-old girls with an osteoblastoma arising from the mandible. In contrast, the oldest patient was an 84-year-old male with an osteoma arising from the mandibular premolar region. The most common decades were 10's and 20's for males and 30's for females.

## DISCUSSION

Lesions involving hard tissue formation are comparatively rare and are generally called fibro-osseous lesions<sup>7,8,11,15,22,23,29-31,33</sup>. In our departments, 1.7% of the lesions were of this

Table 3 Frequency in each region

Lesions	Maxilla			Mandibular			Others	Total
	Incisor	Premolar	Molar	Incisor	Premolar	Molar		
Osteoma	15	31	59	15	38	70	29	257
Osteo-chondroma	0	1	1	0	0	0	2	4
Chondroma	0	1	0	0	0	0	0	1
Osteoid osteoma	0	1	1	0	1	2	0	5
Osteoblastoma	0	0	0	1	3	2	1	7
Ossifying fibroma	13	12	9	15	22	33	1	105
Cemento-ossifying fibroma	2	4	0	4	5	13	0	28
Cementifying fibroma	6	9	9	7	17	30	0	78
Cementoma	4	2	6	2	8	21	0	43
Cementoblastoma	0	0	0	0	0	3	0	3
Gigantiform cementoma	1	3	2	1	3	2	0	12
Periapical cemental dysplasia	1	3	7	0	5	8	0	24
Osteosarcoma	4	3	4	3	7	10	1	32
Chondrosarcoma	1	0	1	0	0	1	0	3
Fibro-osseous lesion	4	7	7	6	18	24	1	67
Fibrous dysplasia of bone	13	12	13	7	16	26	6	93
Total	64	89	119	61	143	245	41	762
Percentage	8.4	11.7	15.6	8.0	18.8	32.2	5.4	100.0

Table 4 Frequency by age on male

	0-9	10-19	20-29	30-39	40-49	50-59	60-69	70-79	80-89	Unknown	Total
Osteoma	2	9	11	10	13	15	16	10	2	2	90
Osteo-chondroma	0	0	1	1	0	0	0	0	0	0	2
Chondroma	0	0	1	0	0	0	0	0	0	0	1
Osteoid osteoma	0	0	0	0	0	1	0	0	0	1	2
Osteoblastoma	1	1	0	0	0	0	0	0	0	0	2
Ossifying fibroma	1	7	3	3	3	2	1	0	0	0	20
Cement-ossifying fibroma	0	2	0	0	1	0	0	0	0	0	3
Cementifying fibroma	0	4	3	4	1	0	0	0	0	0	12
Cementoma	0	4	2	4	4	2	1	1	0	0	18
Cementoblastoma	0	0	1	0	0	0	0	0	0	0	1
Gigantform cementoma	0	0	0	0	0	2	0	0	0	0	2
Periapical cemental dysplasia	0	0	3	0	2	0	0	0	0	0	5
Osteosarcoma	1	0	3	2	1	1	0	0	0	0	8
Chondrosarcoma	0	0	0	0	1	0	0	0	0	0	1
Fibro-osseous lesion	3	3	2	0	1	3	1	0	0	0	13
Fibrous dysplasia of bone	0	8	8	2	3	0	0	0	0	0	21
Total	8	38	38	26	30	26	19	11	2	3	201
Percentage	4.0	18.9	18.9	12.9	14.9	12.9	9.5	5.5	1.0	1.5	100.0

Table 5 Frequency by age on female

	0-9	10-19	20-29	30-39	40-49	50-59	60-69	70-79	80-89	Unknown	Total
Osteoma	0	6	16	26	29	21	11	3	0	1	113
Osteo-chondroma	0	0	0	0	1	0	1	0	0	0	2
Chondroma	0	0	0	0	0	0	0	0	0	0	0
Osteoid osteoma	0	0	1	0	0	0	0	0	0	0	1
Osteoblastoma	2	0	0	0	0	0	0	0	0	0	2
Ossifying fibroma	2	5	13	10	6	2	2	0	0	0	40
Cement-ossifying fibroma	0	0	3	8	4	5	2	0	0	0	22
Cementifying fibroma	0	2	6	12	9	8	0	0	0	0	37
Cementoma	0	0	4	1	6	4	3	1	0	1	20
Cementoblastoma	0	0	2	0	0	0	0	0	0	0	2
Gigantform cementoma	0	0	0	1	2	1	1	0	0	0	5
Periapical cemental dysplasia	0	0	0	1	5	9	2	0	0	0	17
Osteosarcoma	0	1	1	3	2	0	0	1	0	0	8
Chondrosarcoma	0	0	0	1	0	0	1	0	0	0	2
Fibro-osseous lesion	0	9	9	13	15	3	2	0	0	1	52
Fibrous dysplasia of bone	2	5	9	12	3	1	2	0	0	1	35
Total	6	28	64	88	82	54	27	5	0	4	358
Percentage	1.7	7.8	17.9	24.6	22.9	15.1	7.5	1.4	0.0	1.1	100.0

type among all lesions diagnosed histopathologically. However, a unified system for such diagnoses has not been determined<sup>7,23,33)</sup>, and diagnostic decisions can not be made for fibro-osseous lesions at this moment. There-

fore, we have evaluated statistical data about lesions involving hard tissue formation, and we discuss the relevant literature and the mechanisms of the formation of bone, cartilage, or cementum.

### 1. The frequency of each type of lesion

Although cementifying fibromas are reported to comprise approximately 2% of odontogenic tumors<sup>2,34</sup>, 49 cases (8.8%) were seen in this study. Cemento-ossifying fibromas were classified as osteogenic neoplasms by WHO in 1992<sup>24</sup>. Twenty-five cases of cemento-ossifying fibromas were counted in this study, because of the difficulty of distinguishing between the cementum and bone. The diagnosis of cementifying fibroma and ossifying fibroma generally depends on the quantity of bone or cementum<sup>5</sup>. Gorlin and Goldman<sup>9</sup> reported that periapical cemental dysplasias were the most common among lesions with cement formation, but only 22 cases (3.9%) of periapical cemental dysplasia were seen in this study. This lesion involves dysplastic hypertrophy at the tooth root, and many cases of untreated periapical cemental dysplasia exist, suggesting that many cases are not pathologically diagnosed<sup>4</sup>. Sakota<sup>23</sup> reported 6 cases of cementoblastoma and 84 cases of fibro-osseous lesions, while Higuchi *et al.*<sup>12</sup> reported 5 cases of cementoblastoma among 37 cases of cemental lesions. However, Barnes<sup>3</sup> reported that cementoblastomas are extremely rare, and we also treated only 3 cases of osteoblastoma over the 36 years in our departments. Although gigantiform cementoma is also rare<sup>2</sup>, 7 cases were seen in this study. A diagnosis of cementoma was used in a wide broad sense<sup>27</sup>, and it was difficult to determine a final diagnosis of cementoblastoma, periapical cemental dysplasia or gigantiform cementoma. Dahlin and Johnson<sup>6</sup> reported that 3% of benign osteogenic tumors were osteoblastomas and 11% were osteoid osteomas. In this study, however, both tumors were extremely rare; only 4 cases of osteoblastomas (0.7%), and only 3 cases of osteoid osteomas (0.5%) were seen over the 36 years. Osteosarcomas are reported to comprise 20% of general osteogenic malignant tumors, and approximately 7% of osteosarcomas arise in oral and maxillofacial areas<sup>6,10</sup>. Osteosarcomas are the most common type of mesenchymal malignant tumors. On the other hand, chondrosarcomas comprised 10 to 12%

of the lesions and were not rare<sup>15</sup>, but only 3 cases of chondrosarcoma were found in this study. The ratio of fibrous dysplasia of bone arising in the oral maxillofacial area was reported to be 10 to 15% of those in the entire body<sup>10</sup>. The ratio of fibrous dysplasia of bone in this study was 10.0% of all the lesions involving hard tissue formation, which is comparatively high.

### 2. The frequency by gender

Ossifying fibromas, cementifying fibromas, cementoblastomas, periapical cemental dysplasias and gigantiform cementomas are reported to arise in females more commonly than in males<sup>8,19,23,32,33</sup>. This study revealed similar numbers except for cementoblastomas, cementomas, and cemento-ossifying fibromas. Although many reports noted that osteomas, osteo-chondromas, chondromas, osteoid osteomas and osteoblastomas were more common in males than in females<sup>3,15,16,21</sup>, most of these neoplasms arose at the same frequency in females and males in this study. There were only 3 cases of chondrosarcoma in this study; chondrosarcoma generally arise in males more commonly than in females. The ratio of fibrous dysplasia of bone arising in females is reported to be 1.5 to 2.0 times higher than in males, which agrees with our data.

### 3. The frequency in each region

It is known that ossifying fibromas, cementifying fibromas, cementoblastomas and periapical cemental dysplasias arising in the mandibular areas are more frequent than those in the maxilla<sup>8,23,26,33</sup>. This study revealed that the frequency of these lesions arising in the mandibular area was correct except for periapical cemental dysplasias; we found 10 cases of maxillar and only one case of mandibular region. For osteomas, Spjut *et al.*<sup>25</sup> and Barnes<sup>3</sup> reported that the rate arising in maxillar areas is higher than in mandibular areas, but Gorlin and Goldman<sup>9</sup> reached the opposite conclusion. In our study, the frequency of osteomas arising in the maxillar region was the same as in the mandibular region. Ishikawa<sup>15</sup> and Dahlin and Johnson<sup>6</sup> reported

that osteosarcomas arising in the oral and maxillofacial areas were extremely rare, but we noted 2 such cases in the maxilla. In spite of the 38 cases of maxilla and 49 cases of mandibular involving fibrous dysplasia of the bone in this study, it usually arises more frequently in the maxilla than in mandibular regions<sup>3,8,23,29,33</sup>. Although ossifying fibromas and cementifying fibromas arise in the mandibular molar region and fibrous dysplasias of the bone often appear in the maxillary molar region<sup>3,8,23,33</sup>, our statistical data revealed these lesions were more frequent in the mandibular molar region. Ishikawa<sup>15</sup> reported that osteochondromas and chondromas often appeared in the mandibular condyle and coronoideus, but our subjects included only 1 case of osteochondroma in the mandibular condyle.

#### 4. The frequency by age

Ushigome *et al.*<sup>29</sup> reported that the mean age of patients with lesions involving hard tissue formation was 29.5 years old, and Gorlin and Goldman<sup>10</sup> said that such lesions commonly arise in patients over 40 years of age. The average age in our data was 40.1 years old, and this is the same as earlier reports. Although Ishikawa<sup>15</sup> reported that osteochondromas and chondromas arose during the prime of life, the mean age of patients with osteochondroma in this study was 47.5 years old; only 1 case of chondroma arose in a 27-year-old male. Sakota<sup>23</sup> reported that the mean age of patients with ossifying fibromas was 29.5 years old, but other reports stated they arose in young patients up to their prime of adulthood. This study also revealed a mean age for such neoplasms of 30.4 years old. Cementifying fibromas commonly arise in patients around 30 years old<sup>3,15,23,33</sup>, and the distribution of ages of patients with those tumors centered at about 30 years old. It has been reported that the mean age of patients with cementoblastomas was 22.5 years old and that 73% of patients were less than 25 years old<sup>1</sup>. Our 3 cases of cementoblastomas were also in patients 22, 27, and 28 years old. Periapical cemental dysplasias are reported to arise in 20 to 50-year-old females; the average

age is 44 years old<sup>4,15,17</sup>. These cases were distributed in patients from 20 to 60 years old, and the average age was 43.4 years old, which was similar to the earlier published data. On the other hand, osteoblastomas normally arise in patients less than 30 years old<sup>3,13,20</sup>, but, in this study, all cases of osteoblastomas arose in patients under 20 years old, and the mean age was 8.5 years old. The mean age of osteosarcomas arising in the oral area was about 10 years older than those that arose in the general body<sup>15,17</sup>. In this study, the mean age of patients with osteosarcomas was 34 years old, in agreement with the opinion of 10 years older. It is known that the mean age of patients with chondrosarcomas is commonly higher than in those with osteosarcomas<sup>15</sup>. Chondrosarcomas arose in this study in patients 34, 48, and 61 years old. Sakota<sup>23</sup> reported that the mean age of patients with fibrous dysplasias of the bone was 19.4 years, and Ushigome *et al.*<sup>29</sup> reported it as 23.6 years old. However, our data had a mean of 28.1 years old, a little higher than the earlier data, but Barnes<sup>3</sup> and Waldron *et al.*<sup>30,31</sup> reported that fibrous dysplasias commonly arise in patients less than 30 years old.

#### 5. Lesions involving bone, cartilage, and cementum formation

Cells forming bone can be divided into three types in the oral area: osteoblasts lying on the bone surface, osteogenic fibroblasts which have differentiated on the gene level in the periodontal ligament, pulp, and periosteum, and fibroblasts that are stimulated to become osteogenic cells after degeneration. Although osteoblasts are normally present in the surrounding bone surface as resting cells, they become active due to impediments or inflammatory stimuli; then these cells form bone tissue<sup>28</sup>. Osteogenic cells associated with a stimulus also differentiate into osteoblasts. Reactive bone formation is produced in limited areas, but osteoblasts that have changed as a neoplasm proliferate and produce hard tissues indefinitely. Osteomas have been classified as neoplasms in the WHO classification, but osteomas arising around the oral area are



known to be reactive, depending on whether they are due to continuous stimulation or inflammation<sup>13,24</sup>. On the other hand, cementum can be produced by osteogenic cells in the periodontal ligament<sup>14</sup>. Lesions with cementum were classified in the old classification of WHO in 1971<sup>21</sup>, but, in the new classification, cementoblastomas and cementomas are classified as benign cementoblastomas, and periapical cemental dysplasias are classified as non-neoplastic bone lesions<sup>18</sup>.

In conclusion, lesions involving hard tissue formation are thought to be related to osteoblasts or osteogenic fibroblasts, but lesions originating from non-specific fibroblasts cannot produce hard tissues except for calcareous degeneration or myositis ossificans.

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